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A Rare Type of Polydactylous Thumb and its Surgery*)

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ABSTRACT

Resently the authors had the opportunity of performing surgery of a trifurcated thumb, an extremely rare morphological polydactylous condition of the thumb.

Trifurcated thumb had not been included under classification of polydactylous thumb heretofore. Comments were made on this entity from the standpoint of its morphology and consideration was given to its developmental mechanism. Also as surgery was performed with the intention of creating one thumb out of three polydactylous components, discussion was made of the procedure employed.

INTRODUCTION

Polydactyly of the thumb is the congenital abnormality of highest frequency, and as practically all are subject to surgery, its surgical technique assumes as important position in the field of surgery of the hand.

Recently, the authors experienced a case of polydactylous thumb with an extremely rare this case offered many suggestions regarding the overall pathological condition of polydactylous thumb and its treatment, these will be reported together with some considerations.

CASE

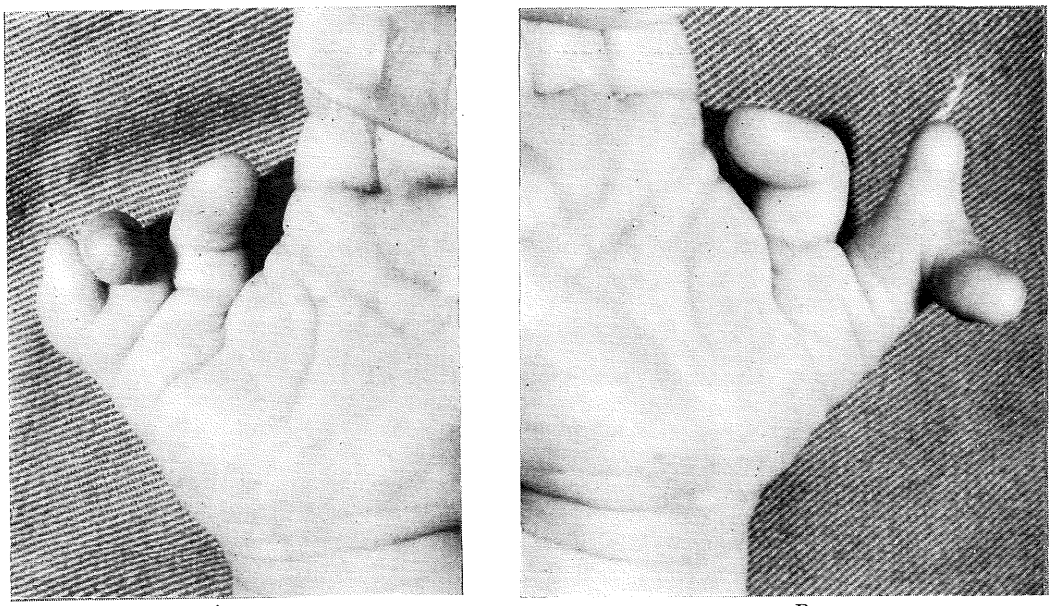
The patient was a boy born on November 22, 1980 as the second son of a 34 year-old father and 30 year-old mother. There was no consanguinity, and the first son, 7, has no abnormalities. There is no remarkable family history.

The mother developed urocystitis during the early part of the second month of gestation and received chemotherapy. At this time, she also suffered severe episodes of morning sickness. She received intravenous infusions but the type of drugs used are unknown. The delivery was uneventful at full term, and birthweight was 3, 400 g. Malfomation of both thumbs was noted immediately after birth, and he was brought to our department with this as the chief complaint, one month after birth. He was admitted to our hospital 9 months after birth for the purpose of surgery.

The external findings at time of admission are shown in Fig. 1-A, B and X-ray fingings are reproduced in Fig. 2-A, B. Both hands presented the extremely rare findings of trifurcated thumb.

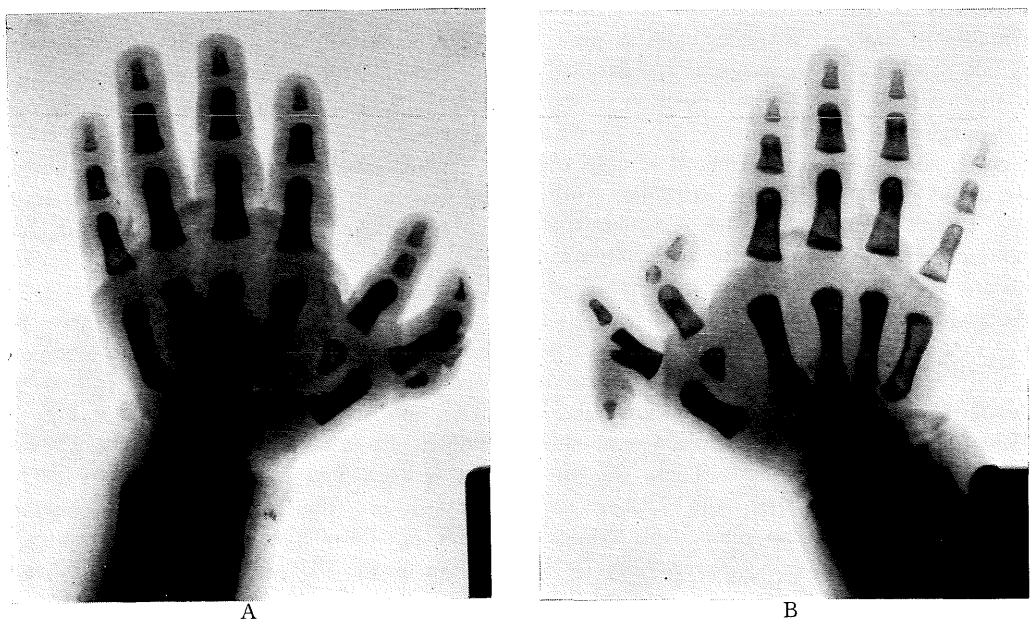
The right thumb consisted of three polydactylous components. The ulnar component was located midway between the first interdigital space and showed the best growth, but morphologically it was a finger and X-ray revealed that it consisited of three phalanges of bony composition. The digital bone corresponding to the middle phalanx was a delta-phalanx, and as a result the DIP joint showed ulnar deviation. Only the head of the metacarpal was retained. To the radial side was located what was considered to be the original thumb, on which was located a single proximal phalanx, but the distal portion of the proximal phalanx divided into two forming the middle poly-

*) 渡　捷一，山口利仁，村岡　博：稀な形態を示した母指多指症とその手術



A B

Fig. 1. Preoperative appearance is shown.
A is the left and B, the right hand. It is the common characteristic findinds that the ulnar component shows the best growth and appearance among three components, and also it located at the center of the first interdigital space. In the left hand, the central and radial components have their own nails but in the right, the central has no nail and the fingertip shows the appearance of transverse terminal arrest. The radial component is floating.



A B

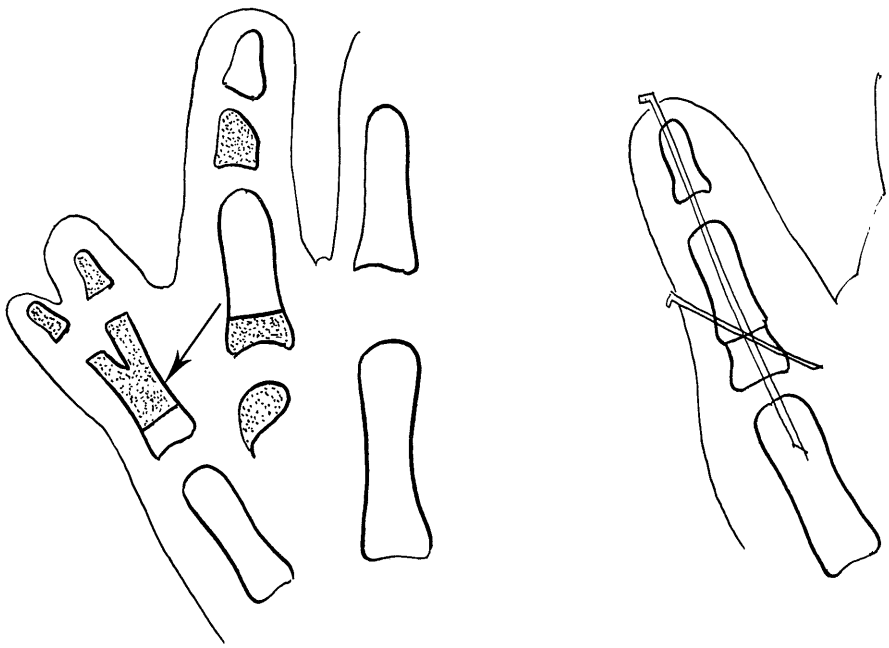
Fig. 2. Preoperative X-ray finding is shown
A is the left and B, the right. The ulnar component of both hands has a rudimental metacarpal and seems to have three phalanges morphologically. On the right hand, middle phalanx is delta-phalanx and DIP joint shows ulnar deviation, but on the left hand, it is rectanglar and no axial deviation is obersved. It is considered that the distal phalanx of the central component of the right was lost by transverse terminal arrest. It had three phalanges originally same as the left.

dactylous component and radial polydactylous component. The middle component had one more phalanx, but it was not immediately possible ascertain as to whether it was a distal phalanx or middle phalanx, but by careful observation, it was noted that the fingertip was slightly round in shape and there was a nail defect which made us assumed that this was the transverse developmental arrest end. Thus, we identified this phalanx to be the middle phalanx. That is, the middle polydactylous component originally had three phalanges. The radial polydactylous component had the poorest growth and was highly unstable. There was a trace of nail on the tip of the digit, and the radiological gap between the distal and proximal phalanges was considered due to disturbance in formation of the middle phalanx. That is, it is presumed that all three polydactylous components had three phalanges.

In the left hand, the ulnar polydactylous component was located midway between the first interdigital space and showed the best growth. Morphologically, it is a triphalangeal digit but different from the right hand, the middle phalanx was rectangular, and ulnar deviation of the DIP joint was not marked. There were three phalanges on top of the first metacarpal, and growth was fairly good. However, growth of the radial polydactylous component was poor and showed two phalanges bifurcating from MP joint.

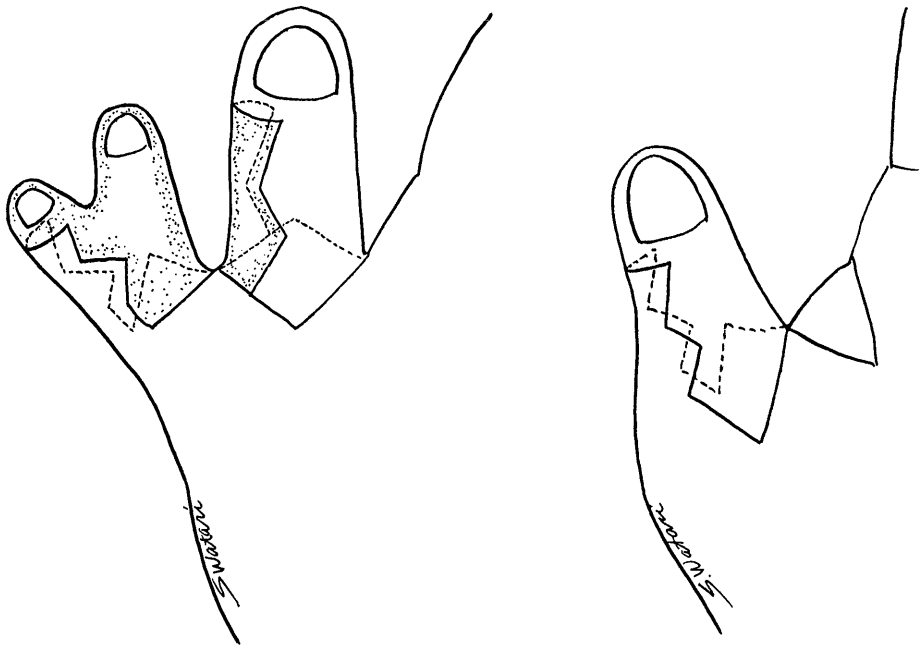
Of the polydactylous components of the left and right hands, those which were mobile were PIP joints of the ulnar component and the MP joints of the middle components.

Surgery in this case was performed from the right hand, but from the cosmetic and functional view points, it was necessary to use as the main source material, the ulnar component which had the best growth and shape. Thus, it become necessary to make tranformation of



A B

Fig. 3. Schematic drawing of the operative procedure
1) Treatment of the bony structure
A : Dotted area shows the portion should be resected.
That is, the ular component of which middle phalanx is removed and osteotomy is performed at the level of the base of proximal phalanx, then transfered to the proximal phalanx of the central component.
B : All procedures are completed.



A B

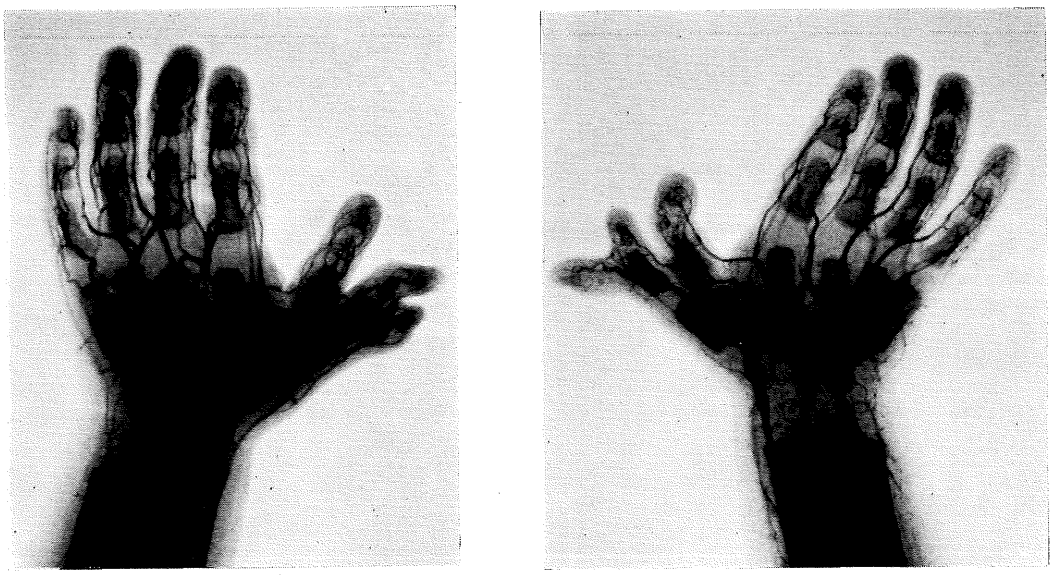
Fig. 4. Schematic drawing of the operative procedure
2) Skin incision
A : Dotted area shows the portion should be resected.
The skin of the ulnar component is incised circumferencially for digital transfer. Fillet made from the radial component is used for the reconstruction of buldging on the radial thumbtip.
B : All procedures are completed.

morphology from finger to thumb by transfer of this component to the thumb position and adjust the number of phalanges and its size[3]. The surgical procedure which consisted of the following was developed, i. e. middle phalanx of the ulnar component was removed and digital transfer to the thumb position of the middle component was accomplished, after which adjustment of thickness was made by skin and soft tissue of the radial component as a fillet.

Fig. 3-A, B show the procedures used on the bony structure, while Fig. 4-A, B show the skin incisions made. The former shows the ulnar component is excised at the base of the proximal phalanx, the extensor tendon, flexor tendon and neurovascular bundle are isolated as a pedicle, and digital transfer of this is made to the proximal phalanx of the middle component. At the same time, the middle phalanx is excised and the distal phalanx is pushed back so as to form the DIP joint. The latter procedure was developed so that adequate exploration could be obtained, the digital transfer could be safely accomplished and adjustment of thickness of the ulnar component could be carried out at the same time.

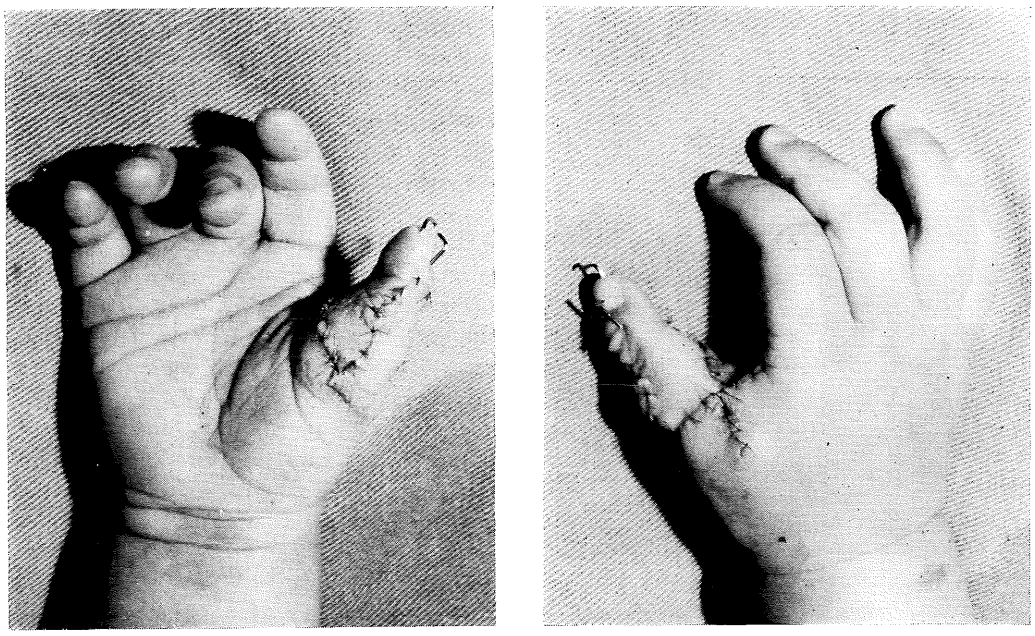
Prior to the actual operation, arteriography was performed to visualize the vasculature ao as to ensure safety of the surgical procedures (Fig. 5-A, B). Fig. 5-A shows the findings of the right hand, and as can be seen although the medial artery is quite dominant, the findings for three arteries, i. e. medial, radial and ulnar, are unique in that there are no communicating vessels linking one another. However, there are branches from the ulnar component distributed to the index finger which is a very interesting finding when it considered that this polydactylous component is derived from the index finger.

Surgery of the right hand was performed on Septemberll, 1981, according to the surgical procedures planned. The postoperative state was satisfactory as shown in Fig. 6-A, B.



A B

Fig. 5. Angiographic findings
A is the vasculature of the left and B, the right hand. There are branches from the ulnar component distributed to the index finger which is a very interesting when it considered that this is derived from the index.



A B

Fig. 6. Findings at the completion of the operation
A : Volar view
B : Dorsal view

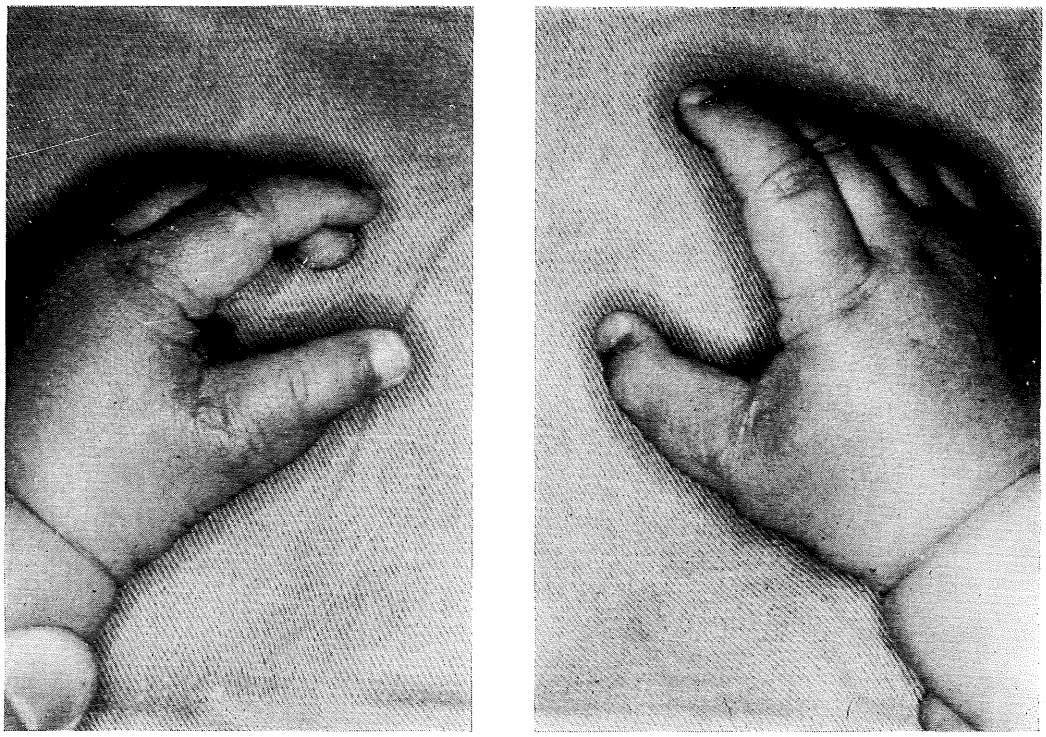
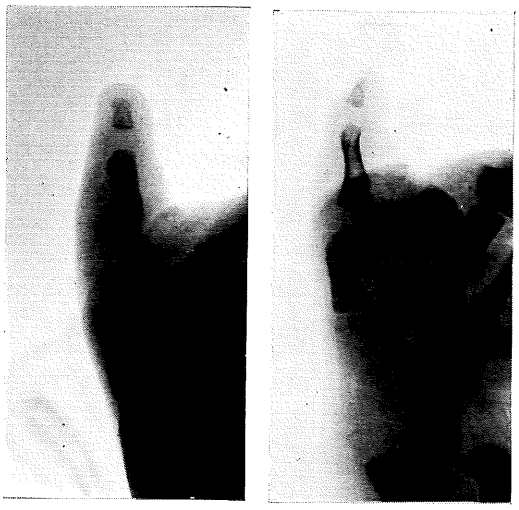


Fig. 7. Postoperative appearance
The right hand shows the state at 6 months after surgery and the left, 3 months after surgery.



A B

Fig. 8. Postoperative X-ray of the right thumb
A : A-P view
B : Lateral view
Digital transfer and arthroplasty of the DIP joint were performed quite satisfactory.

Surgery of the left hand was performed three months later using same procedure as that for the right hand. After immobilization for 3 weeks, check was made of bone union of the digital tranfered site, Kirschner's wires were removed and active exercise was commenced.

Fig. 7 shows the state of the right hand at 6 months and the left hand 3 months after surgery. Although there are slight motor disturbances of the MP and IP joints of both thumbs, they are both very stable and cosmetically acceptable. Of course, there is no difficulty in carrying on daily life activity.

Fig. 8-A, B are X-ray films of the right thumb. The bony structure of the thumb is well formed and congruency of the IP joint is good. No axial deviation is observed.

DISCUSSION

1. Morphology of polydactylous thumb

The morphological classification of polydactylous thumb most widely used is that of Wassel[4]. This is shown in Fig. 9. There is no arguing that the most highly evaluated point in this classification is the ascertainment of the

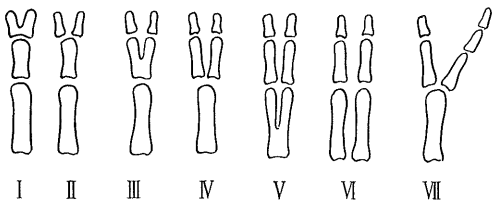


Fig. 9. Wassel's classification
Polydactylous components in types I-VI have two pahalanges respectively but in type VII either or both components have three phalanges.

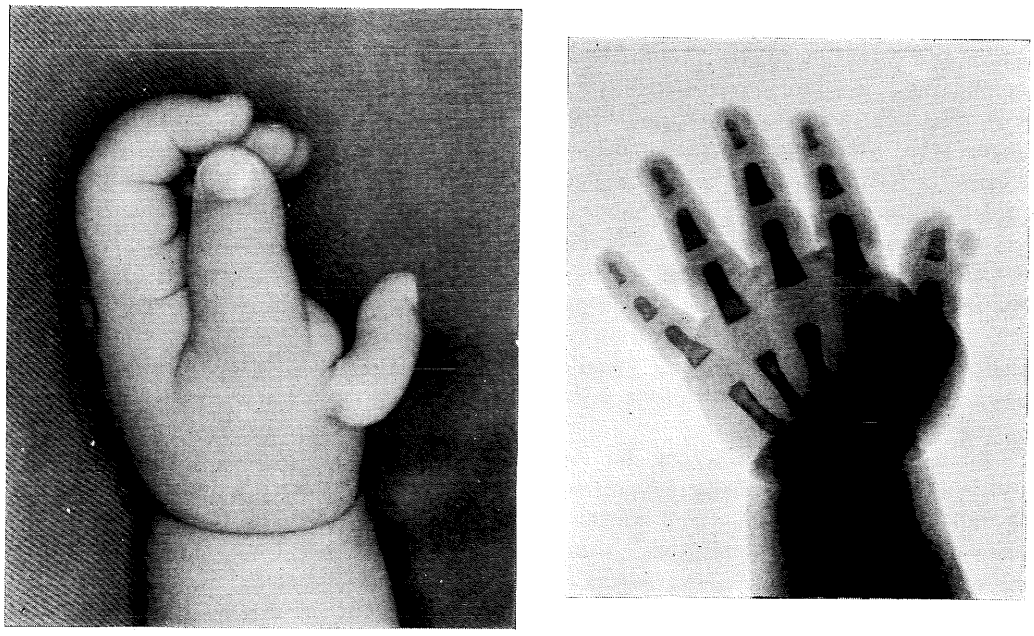
presence of type VII, but there are also not a few points which have caused claims of inadequency and misunderstanding.

First, there is a problem which is common to all classification of the polydactylous thumb and must always be borne in mind. That is, the X-ray findings of this entity can be observed at the appropriate age for surgery, up to about age 1, but no one knows about the state thereafter, particularly the permanent image after bone growth has been completed. In other words, the X-ray image we are observing routinely, is a transient form and nothing definite can yet be said. One should be careful and also flexible in determining the number of phalanges and identifying the phalanges.

Therefore, all conditions in which bifurcation occurs at the distal phalanx immediately after bith are type II while the proportion of those with type I which increases with time is merely because ossifications of cartilages do not take place, thus there is not such a sereous meaning in the classification of types I to VI. It is felt types I and II may be called distal phalangeal type, types III and IV the proximal phalangeal type and types V and VI the metacarpal type.

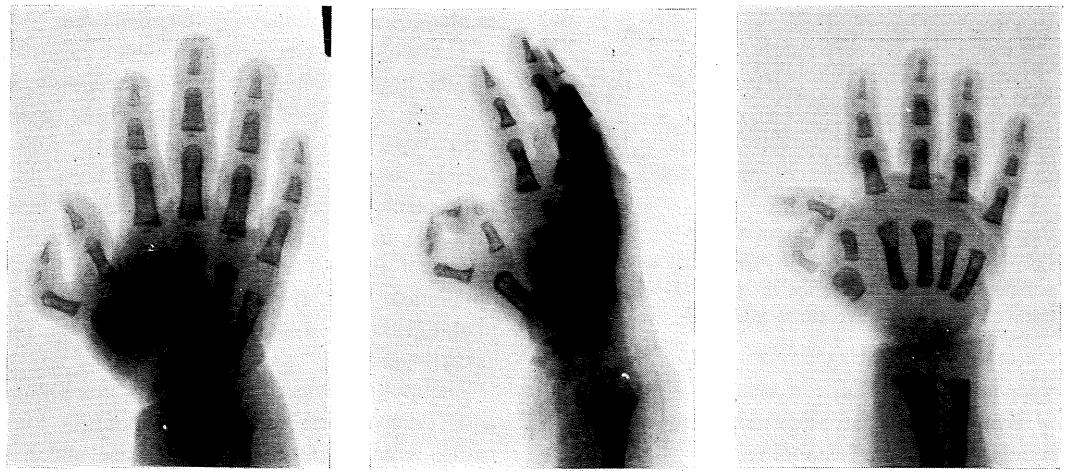
The polydactylous components are either of equal size or the radial component is hypoplastic, which is more often the case and if this continues to progress it becomes rudimentary making it impossible to maintain a close relationship with the digital bony component and joint. In extreme cases it is reduced to a wart-size or is attached as merely a floating digit. Such condition are not rare, and we, aside from Wassel's classification, have grouped these under rudimentary type. It is felt an independent category should be established for such polydactylous thumb from the standpoint of morphology and choice of treatment (Fig. 10).

One of the polydactylous components in type



A B

Fig. 10. Rudimentary type of polydactylous thumb
Usually, polydactylous components are either of equal sive or the radial is hypoplastic. In extreme cases it is reduced to a wart-size (A) or is attached as merely a floating digit (B).



A B C

Fig. 11. Various morphology of the cases in type VII
There are three types in the proximal phalangeal type; only radial component shows three phalanges (A), Both components show three phalanges (B), only ulnar component shows three phalanges (C).
Bifurcating level also varies from metacarpal type to distal phalangeal type.

VII is said to have a triphalangeal component and the schematic drawing presented a proximal phalangeal type polydactyly in the original article[4]. However, it is well known that the proximal phalangeal type of type VII consists of three subtypes, i. e. ulnar component with three phalanges, radial component with three phalanges and three phalanges in both of the two components. Further, in polydactylous type there is also the metacarpal type, and distal phalangeal type. Also when there is hypoplasia, disturbance in formation of the middle phalanx is noted like brachysyndactyly, and at time of surgery there are many in whom the ossification center has not yet appeared resulting in a large gap between the distal and proximal phalanges. It is felt that if surgery were not performed on such cases, the proportion of those of so-called type VII polydactylous thumb would be markedly greater (Fig. 11).

Of course, there is no category for our trifucated thumb case. Generally as polydactylous thumb implies bifurcation and is defined as a two division of the thumb, such a trifurcation is an exception. The oldest report of such a polydactylous morphology is felt to be that by Desnoyers in 1924[2]. We have been able to locate few reports of such cases in recent literature[5], and this is the first in which the surgical experience is discussed. We have in the past experienced an incomplete type trifurcated thumb, but it is felt a completely separated type such as this is extremely rare. However, in our experimental studies to induce malformation of the extremities of mice using 5-FU, this type of malformation was rather frequently encountered, and the finding observed revealed that all three polydactylous components were triphalangeal is of great interest when comparison is made aginst our case. Further, as it was strongly suspected that polydactylous components of the index finger were affecting the thumb, this finding has great implications when considering the developmental mechanism of polydastyly of the thumb.

2. Surgical policy for polydactyly of the thumb

The history of surgical treatment of polydactyly of the thumb is not well known but it is considered that the polydactylous component was excised immediately after birth as being excessive. As polydactyly of the thumb means there are two thumbs, the concept until very recently had been to eliminate the excessive component and match the number of digits, but treatment of the secondary or remnant deformity which always occured after such operation posed a problem greater if not equal to that of untreated cases. This is evident from the results of our twenty-year experience

in hand surgery whereby postoperative deformity in polydactylous thumb during the first ten years out-numbered the untreated cases. However, the majority of these operations had been performed by non-specialist, whereas those who specialized in this field were of the opinion that the supernumerary component should not be eliminated, but that of the tissue of the polydactylous components should be collect together to reconstruct one new thumb. The most typical procedure is the Bilhaut-Cloquet[1] methode which is a bisecting and joining method. The procedure originally was used for treatment of the distal phalangeal type, but recently its use has been extended to include the proximal phalangeal type as well. It is used when the polydactylous components are of about the same size and it is considered that if only one component were retained, the thumb would be too slender. This has been called an extended bisecting and joining method and appears to be a logical procedure, but in practice as it is difficult to split two phalanges into two and join them into one, such procedure can be performed only by a limited number of surgeons and thus good results can be achieved in only a limited number of cases. Therefore, generally speaking, the functional and cosmetic prognosis was not good. We were obliged to perform secondary and tertiary corrective procedures of such cases on a number of occasions, and it was often necessary to totally excise one of the fused nails and phalanges. Thus, we began to feel that it would be better to add a filled of the radial component to the ulnar component and enlarge the size and shape of the thumb rather than joining the nails and phalanges by primary surgery. In determining the indication for the extended bisecting and joining method, we had attached a great amount of weight upon the width of the finger nail as compared to the healthy side, but later found that for the patients and their parests the shape of the tip of the thumb was of far greater concern than the nail so long as it was not too unnatural.

In accordance with the above, we have been employing the techniques of the extended bisecting and joining method using only the nail and phalanges of the ulnar component and adjusting the size with a fillet taken from the radial component. Also we have been applying this procedure to situations where the radial component is definitely inferior and would have been simply excised in the past and have succeeded in plasty of the thumb to make it appear more natural.

In other words, it is a mistake to consider polydactyly of the thumb as there being an excessive component and performing a subtraction operation to match the number of digits, it should be thought of as an additive operation where one thumb had been separated into a number of components, and thus necessary tissue from the respective components should be brought together to reconstruct one new thumb. From this point of view, it is felt that the operation performed on this case was symbolic. That is, the ulnar component provided the main bony structure and shape of thumb, the central component provided the seat of thumb, while the radial component, improvement of shape.

ACKNOWLEDGMENT

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